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Case Report

Post-extraction Inflammatory Myofibroblastic Tumor of Maxilla – A rare oral presentation

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Abstract

Inflammatory myofibroblastic tumor (IMT) is a rare, benign, locally aggressive tumor that shows uncertain malignant potential. Based on the spectrum of histological appearances, it is known by different names, most commonly plasma cell granuloma, inflammatory pseudotumor, myofibroblastoma, inflammatory myofibrohistiocytic tumor, xanthogranuloma, fibrous xanthoma, etc., and most recently as IMT. The etiology is non-specific and suggested to be a result of an inflammatory response to unknown factors. It is particularly uncommon in the oral cavity, with a PubMed literature search identifying only two documented cases reported at post-extraction sites to date, with the most common clinical differential diagnosis of peripheral reactive lesions such as pyogenic granuloma, peripheral giant cell granuloma, and peripheral ossifying fibroma. This article presents a rare case of an IMT with an aggressive clinical presentation, arising at the post-extraction site of the right maxilla in a 50-year-old female patient. We also aim to discuss its etiopathogenesis, clinical and radiological features, histopathological correlations, and management strategies. Although uncommon, IMT should be considered in the differential diagnosis of post-extraction lesions in the oral cavity due to its potential for local recurrence, metastasis, and rare malignant transformation. Therefore, prompt diagnosis and sustained, long-term follow-up are essential to ensure favorable clinical outcomes.

Keywords: Inflammatory myofibroblastic tumor, Inflammatory pseudotumor, Oral cavity, Maxilla, Myofibroblast, Smooth muscle actin.

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1. Introduction

Inflammatory myofibroblastic tumor (IMT) is a rare, benign but locally aggressive tumor.[1] It was earlier known as inflammatory pseudotumor, as it presents the characteristics of malignancy, such as expansion, invasiveness, metastasis, and recurrence.[2] This term was later recoined as IMT by Scott et al. in 1988 and was first described as a tumor by Umiker and Iverson.[3] In 1994, WHO considered this condition a distinct entity[4] and defined IMT as "an intermediate soft tissue tumor that is composed of myofibroblast-differentiated spindle cells and accompanied by numerous inflammatory cells, plasma cells, and/or lymphocytes".[2,5] The origin of the tumor is uncertain and shows varied clinical presentations.[6] The most common sites of occurrence are lungs, followed by the abdomen and pelvic region.[7] In the head & neck region, the highest predilection for IMT is in the orbit, whereas, in the oral cavity, it is predominantly seen in the buccal mucosa, mandible, and maxilla in descending order.[8,9] In this article, we have presented a rare case of IMT occurring in the maxilla arising from a post-extraction site. Extensive research on PubMed literature revealed only two cases of IMT arising from the extraction socket that have been reported so far,[4,10] making the present case the 3rd rarest occurrence from the extraction site.

2. Case Presentation

A 50-year-old female patient reported to the outpatient department of Oral Medicine and Radiology in a tertiary care center with a chief complaint of swelling on the right side of the face and gums in the upper right posterior teeth region. The patient had undergone extraction of a maxillary posterior tooth on the right side one year ago. Following the extraction, she developed persistent pain in the same area, which was later accompanied by a gradually enlarging swelling. Over

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time, the swelling progressed, resulting in noticeable facial asymmetry. On extraoral examination, the swelling extended from the right infraorbital region up to the angle of the mouth superoinferiorly and approx. 2 cm in front of the right tragus of the ear posteriorly and up to the right ala and the lateral wall of nose anteriorly with obliteration of the nasolabial fold and drooping of the right angle of the mouth (**Figure 1**). Intraorally, a well-defined exophytic growth, approximately 5 x 4 cm (anteroposterior x buccopalatal) in its greatest dimensions, was present attached to the right buccal vestibule and alveolar ridge of edentulous 16 region, extending anteriorly over the buccal aspect of the 13,14 and 15 regions with surface indentations of the buccal cusps. The surface of the growth was smooth but lobulated on palpation, with few areas of erythema. The growth was associated with a hard, non-tender swelling on the palatal aspect of the 16 and 17 regions with surface ulceration and erythematous margins. The hard tissue examination showed generalized attrition of teeth with sharp cuspal margins of 13,14 and 15 (**Figure 2** and **Figure 3**). On correlating the history and clinical findings, a provisional diagnosis of peripheral giant cell lesion was made with differential diagnoses of peripheral ossifying fibroma, epulis granulomatosum, and soft tissue malignancy.

Contrast-enhanced CT revealed a well-defined, heterogeneously enhancing soft tissue lesion measuring 3.6 x 3.2 x 2.5 cm (TR x CC x AP) arising from the edentulous upper right 1st molar region (**Figure 4 A** and **B**). It extended medially, eroding the buccal and palatal cortices of the superior alveolar arch into the oral cavity. Laterally, it extended into the ipsilateral gingivobuccal space with lateral displacement of the depressor anguli oris muscle; however, the fat planes remained intact. Superiorly, the lesion caused thinning of the floor of the right maxillary sinus and the right lateral aspect of the palate. Inferiorly, the lesion was abutting the inferior alveolar arch with no obvious bony erosion (**Figure 5**).

FNAC performed from the buccal aspect of the growth revealed only blood, and the sample obtained from the palatal swelling yielded a cellular smear that consisted of plump, oval-to-spindle cells dispersed in an abundant fibromyxoid background admixed with inflammatory cells (polymorphs and plasma cells). Individual cells showed mild nuclear polymorphism. The cytological features were in favor of a benign myofibrohistiocytic/ myofibroblastic tumor.

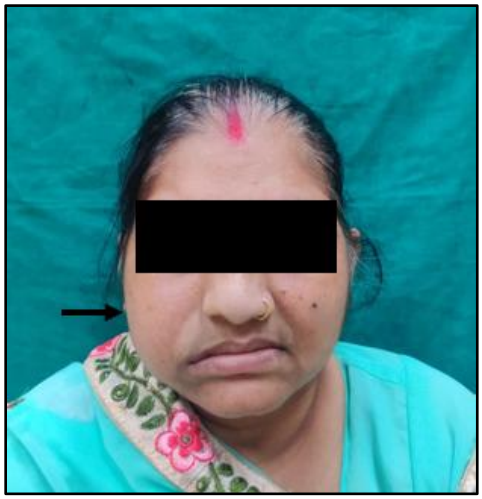

Figure 1: Extraoral photograph showing diffuse swelling on right cheek region (black arrow) with drooping of right angle of mouth.

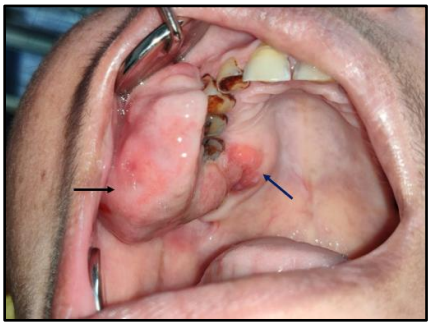

Figure 2: Intraoral photograph showing a well-defined growth arising from edentulous 16 region and extending buccally and anteriorly (black arrow). Presence of palatal swelling with surface ulceration and erythema (blue arrow).

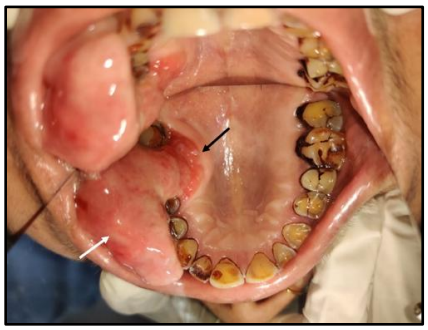

Figure 3: Intraoral occlusal view of maxilla showing the site of origin of the growth from partially edentulous 16 region, extending buccally and anteriorly (white arrow) and associated palatal swelling (black arrow). Generalized attrition of teeth noted with brown-black stains.

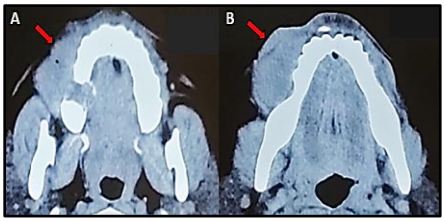

Figure 4: A and **B.** Axial sections of contrast enhanced CT (CECT) showing a well-defined heterogeneously enhancing soft tissue lesion in right posterior maxilla arising from edentulous 1st molar region with erosion of buccal and palatal cortices (red arrows).

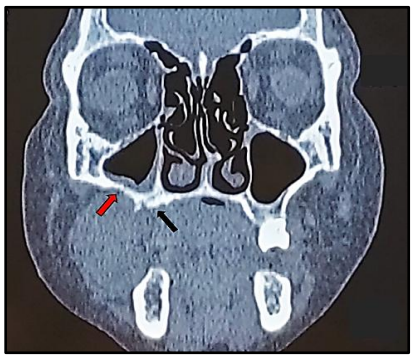

Figure 5: Coronal section of CT showing an expansile lesion in the right oral cavity causing thinning of the floor of the right maxillary sinus (red arrow). Associated mucosal thickening is noted along the sinus floor and medial wall. Erosion of buccal and palatal cortices of upper alveolar arch (black arrow).

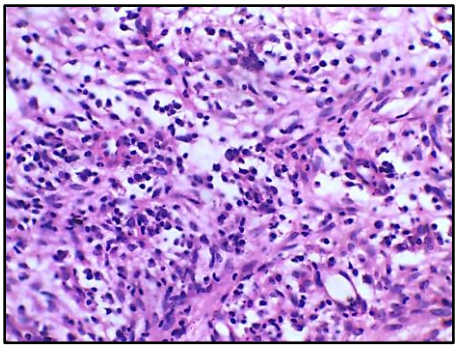

Figure 6: Photomicrograph showing a cellular tumor with spindle shaped cells intermingling with plasma cells, lymphocytes and eosinophils in a loosely collagenous background. (H&E X 40)

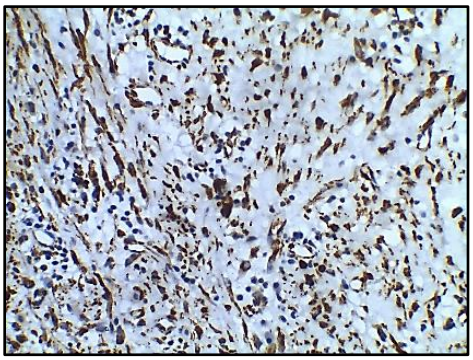

Figure 7: Photomicrograph showing immunoreactivity of spindle cells for smooth muscle actin (SMA) (X 40)

Table 1: Review of literature of cases of inflammatory myofibroblastic tumor of jaws arising from extraction site

Author	Age	Gender	History of extraction	Site	Clinical features	Treatment	Follow-up	Recurrence at follow-up
Biniraj KR et al[10]	38 years	Female	2 months ago	Left maxilla; 27 region	Non-tender swelling; 3 x 4 cm in diameter	Left side maxillectomy followed by radiotherapy	6 months	Yes, finally succumbed to death
Eley KA et al[14]	29 years	Male	1 month ago	Right maxilla; 15 region	Large, tender swelling; 5 cm in diameter	Surgical resection	6 years	No
Ramasamy P et al (present case)	50 years	Female	1 year ago	Right maxilla; 16 region	Large, non-tender growth; 5 x 4 cm in diameter	1-week pre-op corticosteroid injection followed by surgical resection	1 year	No

Incisional biopsy was performed under local anesthesia, and two deep soft tissue samples were obtained, one from the buccal aspect and another from the palatal swelling. Histopathology revealed loose collagenous fibrocellular connective tissue stroma with numerous uniform spindle cells containing vesicular nuclei with pale eosinophilic cytoplasm. Mild nuclear pleomorphism was observed. Diffuse dense inflammatory infiltrates chiefly composed of plasma cells, lymphocytes, neutrophils, and eosinophils were evident. Varying-sized blood vessels with perivascular lymphocytic infiltration were noted. The ulcerated surface of the lesion was covered with fibrin and enmeshed with inflammatory cells (**Figure 6**). Immunohistochemical analysis revealed strong smooth muscle actin (SMA) expression, confirming their myofibroblastic phenotype (**Figure 7**). Thus, a final diagnosis of IMT was made. Notably, the tumor was negative for ALK expression, a feature that highlights the heterogeneity of IMT. A whole-

body PET scan demonstrated no signs of distant metastatic spread, suggesting localization of IMT to the primary region.

The treatment provided for the present case was a 1-week corticosteroid injection followed by surgical excision. There were no intra-op and immediate post-op complications. The patient remained asymptomatic at the 1-year follow-up conducted through telephonic consultation, although no in-person evaluation was performed.

3. Discussion

Inflammatory myofibroblastic tumor (IMT) is a spectrum of rare benign soft tissue neoplasms expressing various characteristics ranging from reactive to malignant presentations.[10] This tumor is known by different terms, namely, plasma cell granulomas, fibrous histiocytoma, benign myofibroblastoma, xanthomatous granuloma, inflammatory pseudotumor, pseudosarcomatous myofibroblastic proliferation, and spindle cell pseudotumor.[11] These reactive lesions were collectively called inflammatory pseudotumor (IPT), and the recent term proposed is IMT, which denotes a neoplasm with low malignant potential.

The lung was the first organ observed to be affected by IMT by Brunn in 1939. It can occur in other organs as well, the most common being the abdominopelvic tissues. IMT rarely occurs in the head and neck region, constituting only about 14% of all cases,[10] with a predilection for the orbit and paranasal sinuses.[12] It also has been reported in salivary glands, the thyroid gland, tonsils, and the trachea. In the oral cavity, the highest predilection is for buccal mucosa, with the mandible being the next most common.[11] Our patient had an intraoral growth, mimicking a malignant lesion, arising from the extraction site of the maxilla, representing a rare entity.

The etiology of the tumor is poorly understood despite various proposed concepts. Tissue trauma followed by aberrant immune response is considered the most acceptable etiological concept. IMT represents an immune reaction to injurious & infectious stimuli such as microorganisms, tissue damage, foreign bodies, or even neoplastic tissues.[13,14] In the present case, trauma due to extraction could have stimulated a chronic inflammatory reaction that resulted in mesenchymal cell proliferation as a tissue response.

The role of myofibroblasts in granulation tissue formation & wound healing suggests IMT to be a direct reactive nature of the lesion.

Biniraj et al[10] reported a case of a 38-year-old female with IMT of the extraction site of left maxilla. The extraction of the maxillary 2nd molar was performed 2 months earlier. Eley KA et al.[4] reported a case of a 29-year-old man with a history of extraction of the right maxillary second premolar 1 month earlier, after which he developed swelling over the extraction site and alveolus, which showed further increase in size.

More recently, a neoplastic origin has been suggested that challenges IMT being recognized as a benign entity with an increased rate of recurrence. The gene responsible for the pathogenesis of IMT is the ALK gene localized to the chromosome band 2p23, which supports the neoplastic origin of the tumor.[15] Radiation exposure or radiotherapy is also considered to have a role in the onset of IMT.[9,16]

IMT may occur at any age, although it preferentially occurs in children and young adolescents, whereas the present case has been observed in the 5th decade of life.[17] Proliferation of mesenchymal cells following an inflammatory stimulus is observed in elderly patients, whereas pediatric IMTs are associated with chromosomal abnormalities, including ALK gene rearrangement.[7]

IMT shows a non-specific clinical picture, varying from symptomatic lesion to well-defined swelling or outgrowth that shows slow progression over years. Clinically, the tumor can be very large and aggressive and may mimic a malignant lesion with invasive and metastatic potential.[9] Lazaridou M et al[13] presented a large lesion of the maxilla occupying the left maxillary sinus, infiltrating the left zygomatic bone, floor of the orbit and nasal septum. The clinical differential diagnosis of oral IMTs includes peripheral giant cell granuloma, pyogenic granuloma, peripheral ossifying fibroma, and soft tissue malignancies.

Radiographically, a mildly enhancing soft-tissue mass without any internal calcification or bone destruction can be seen. As a mesenchymal tumor of soft tissue, the tendency for the tumor to cause underlying bone erosion and destruction is present, which favors the high suspicion of a malignant tumor.[11] Resorption of multiple teeth and the perforation of the palatal cortex are additional features of local aggression.[15] CT and MRI are the imaging modalities of choice to identify the location and extension of the lesion. The radiographic differential diagnosis includes aggressive odontogenic tumors, mesenchymal odontogenic tumors, soft tissue malignancies, fibrosarcoma, and low-grade myofibrosarcoma.[10]

In MRI, the enhancement mode and signal intensity tend to vary based on the histological composition of the tumor. If the borders appear well-defined with peritumoral edema, it represents a morphologically benign condition. In case the ratio of mucus component is high, T1-weighted images show iso- or slight hyperintensity, and T2-weighted images show bright hyperintensity. Hypointensity in both T1- and T2-weighted images constitutes predominantly collagen fibers. Based on the contrast enhancement, the aggressiveness of the lesion can also be correlated with the MRI features. Indolent tumors appear hypointense on T2-weighted images with no enhancement, whereas fast-growing and aggressive lesions appear hyperintense on T2-weighted images with marked contrast enhancement.[7]

Histopathological examination along with adjunct tools like immunohistochemistry is necessary for establishing an accurate diagnosis.[6] In IMT, myofibroblasts are mesenchymal cells with intermediate characteristics between fibroblasts and smooth muscle cells.[18] There are 3 basic histological variants of IMT: inflammatory, cellular, and few-cell type.[12]

1. Inflammatory type - loosely organized myofibroblasts in an edematous myxoid background with plasma cells, lymphocytes, eosinophils, and blood vessels, resembling nodular fasciitis, are present.
2. Cellular type - dense aggregates of spindle cells arrayed in a variable myxoid and collagenized background; admixed with a distinctive inflammatory infiltrate (plasma cells, lymphoid nodules) resembling fibrous histiocytoma or fibromatosis. Our case presented features similar to the cellular type.
3. Few-cell type - collagen sheets with scattered plasma cells and eosinophils resembling a scar or desmoid tumor.

The differentials for histologically benign tumors include nodular fasciitis, solitary fibrous tumor, desmoplastic fibroma, benign fibrous histiocytoma, myofibroma, calcifying fibrous tumor, and plasmacytoma. Histologically, malignant features resemble low-grade fibromyxoid sarcoma, myxofibrosarcoma, myofibrosarcoma, follicular dendritic cell sarcoma, leiomyosarcoma, fibrosarcoma, anaplastic large cell lymphoma (ALCL), malignant peripheral nerve sheath tumor (MPNST), and spindle cell carcinoma.[19]

Immunohistochemistry may reveal positivity for SMA, vimentin, muscle-specific actin, cytokeratin, and anaplastic lymphoma kinase (ALK). Approximately 50% of IMTs show a strong association with ALK positivity, which indicates the aggressiveness of the lesion. 0-10% IMT expresses positive nuclear expression of Ki67, which reflects the underlying neoplastic process and is an indicator of ongoing proliferation activity.[19]

The treatment of choice is surgical excision in combination with corticosteroids. It has been stated that around 80% of IMTs respond well to corticosteroids, but not an ideal single-treatment modality due to incomplete remission. High-dose corticosteroids are effective in non-aggressive, non-neoplastic lesions. In case of non-resectable & non-responsive cases and those with ALK-1 & Ki67 positivity, radiotherapy acts as an adjunctive therapy given in the dose range of 50-54 Gy for low risk, 60-64 Gy for moderate risk and 66-70 Gy for high-risk lesions.[20] Postoperative assessment is advised for a minimum of 10 years to look for recurrence.[2]

Metastasis is rare and evident in less than 5% of the cases.[15] Gallego et al[2] reported a case of lung IMT with metastasis to the maxillary region in a 53-year-old female, representing the multifocal nature of IMT with pulmonary and extrapulmonary lesions. The recurrence rate of IMT is 37%. IMTs with ALK positivity, elevated Ki-67 proliferative index (up to 10%), DNA aneuploidy, or oncogenic protein overexpression (p53, bcl-2) exhibit increased risk for recurrence.[2] The malignant transformation in IMT occurs in a subset of cases that often results in fibrosarcoma.[2]

Table 1 provides a review of three known cases of IMT in the oral cavity (maxilla) arising from an extraction site, including ours. The present case highlights its uniqueness through a highly uncommon presentation in terms of patient age, duration, and clinical features arising at a prior tooth extraction site. Remarkably, despite aggressive local progression, no metastasis was observed.

4. Conclusions

IMT is a rare differential diagnosis of proliferative lesions arising from an extraction site with non-specific presentation both clinically and radiographically. Though it is a rare benign tumor, the lesion displays malignant characteristics with a significant rate of recurrence and metastasis. Hence, dental professionals play a crucial role in the diagnosis of oral IMT, correlating the clinical history and presentation, radiographic findings, and histopathological analysis with immunohistochemistry and adhering to the appropriate treatment modalities for quicker recovery. Regular and long-term follow-up is crucial to avoid recurrence and metastasis, thereby reducing the morbidity and the chances of malignant transformation.

5. Source of Funding

None.

6. Conflict of Interest

The authors report no conflicts of interest related to this case report.

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